

Case Report

Duodenal duplication manifested by abdominal pain and bowel obstruction in an adolescent: a case report

Xiaoyu Yan, Ying Fan, Kai Wang, Wei Zhang, Yanglin Song

Department of The Second General Surgery, Shengjing Hospital of China Medical University, Shenyang, Liaoning Province, People's Republic of China

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Abstract: Duodenal duplication (DD) is a rare congenital anomaly reported mainly in infancy and childhood, but seldom in adolescent and adults. Symptoms, such as abdominal pain, nausea, vomiting or dyspepsia may present depending on the location and type of the lesion. DD can result in several complications, including pancreatitis, bowel obstruction, gastrointestinal bleeding, perforation and jaundice. Surgery is still the optimal method for treatment, although endoscopic fenestration has been described recently. Here, we report a case of a DD on the second portion of the duodenum in a 17-year-old adolescent complaining of transient epigastric pain and vomiting after meal. We suspected the diagnosis of DD by abdominal computerized tomography and endoscopic ultrasonography. We treated her by subtotal excision and internal derivation. Eventually, we confirmed our diagnosis with histopathological result.

Keywords: Duodenal duplication, congenital anomaly, duodenum, abdominal pain, bowel obstruction, subtotal excision, internal derivation

Background

Duodenal duplication (DD) is a rare congenital malformation of the gastrointestinal tract [1-10] and accounts for only 2%~12% of all gastrointestinal (GI) tract duplication [5, 6, 8]. It is usually diagnosed in the pediatric population, and only 30% of patients are diagnosed elder than 12 years old. According to the literature, the eldest patient diagnosed as DD was 73 years old [2]. To our knowledge, it is a great challenge for surgeon to diagnose, as its unknown etiology and nonspecific symptoms. We report a rare case of DD with duodenal obstruction presenting as transient epigastric pain and vomiting after meal.

Case presentation

A 17-year-old adolescent was admitted to our hospital complaining of 3-years history of transient epigastric pain and vomiting after meal for previous 20 days. She had no remarkable past medical and familial history. No anomaly was found on physical examination, and all laboratory examination was within normal ranges.

A gastroduodenoscopy revealed a four-centimeter smooth mass in the second part of the duodenum as well as erythema exudative gastritis (**Figure 1**). Upon abdominal contrast-enhanced computerized tomography, a cystic lesion of 3.4×3.3 cm in cross-section was found which located at the second portion of the duodenum (**Figure 2A**). Furthermore, it also demonstrated that the lateral wall of the second part of the duodenum became thicker, and there was restricted liquid between tunica mucosa and tunica serosa of the lesion (**Figure 2B**). To find out the relationship between the cystic lesion and the biliary and pancreatic system, an endoscopic ultrasonography was done, which revealed no communication existed (**Figure 3**). One week later, operation was performed and we found an elliptic cystic lesion of 3.4×3.5 cm in diameter and anterolateral to the second part of the duodenum (**Figure 4**). We performed a paracentesis on the cystic lesion and found faint yellow liquid in it, and no communication between the lesion and duodenum was observed. We performed the subtotal resection and the marsupialization which was also called

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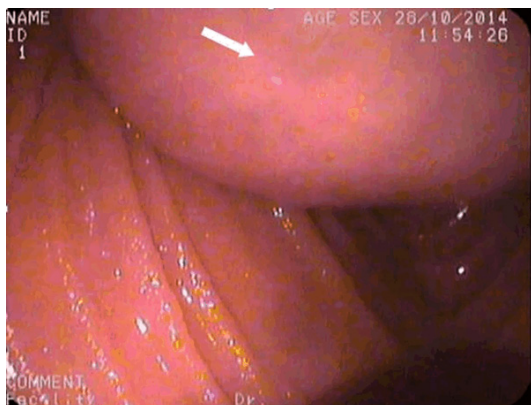


Figure 1. A gastroduodenoscopy revealed a four-centimeter smooth mass (white arrow) in the second part of the duodenum as well as erythema exudative gastritis.

classical Gardner-Hart operation (**Figure 5**). Eventually, the diagnosis was confirmed by histological findings, and duodenum-like mucosa, submucosa and muscle coats were found from the resected tissue (**Figure 6**). The patient's postoperative recovery was uneventful and 5 days later she was discharged home. A abdominal CT was done in 3 months later which resulted for no abnormality (**Figure 7**), and she was asymptomatic after 5 months follow-up.

Discussion

DD is congenital malformation formed during the embryonic period of gastrointestinal tract development [1-10]. However, the duplications of GI occur most commonly in the distal ileum, followed by the esophagus, colon and jejunum [2, 3, 5, 8]. Only 2%~12% of GI duplications are located in the duodenum [5, 6, 8]. DD is usually diagnosed in infancy and childhood. A meta-analysis which included 47 cases of DD found that the number of patients who were in the pediatric period (40.4%) was more than the patients who were elder than 20 (38.3%). Nevertheless, the least number of patients were in the second decade (21.3%) [6]. DD can be divided into tubular or cystic, communicating or non-communicating. Most are located in the first or the second portions of the duodenum [1-3]. DD reported in our case was cystic and non-communicating type, and located in the second part of the duodenum.

The clinical manifestations of DD are variable which are determined by the site, type and size

of the lesions. The most common symptoms are abdominal pain and nausea/vomiting, and the most common complications are pancreatitis, which occurred in 53% of DD [5, 6]. Other manifestation such as gastrointestinal obstruction, infection or intussusception has also been reported. Obstruction is usually presented in non-communicating cystic duplication, as shown in our patient, due to the intracystic secretions making the duplication cyst distended to compress the gastrointestinal tract.

There are 3 criteria for the diagnosis of the DD, including an intimate attachment to the native gastrointestinal tract, a muscle coat, and alimentary mucosal lining [2, 6]. However, the pre-operative diagnosis is still inaccurate due to the nonspecific clinical manifestation and unknown etiology. Imaging modalities can help detect the lesions to make our diagnosis become more accurate [11-15]. Ultrasonography can distinguish the bowl structure of the duodenal duplication from other cystic lesions, and an echogenic inner mucosa surrounded by a hypoechoic outer muscular layer is seen on ultrasonography [14]. We usually use endoscopic ultrasonography to reveal the relationship of the DD and the pancreaticobiliary system, which can differentiate the choledochal cyst, especially Todani type III. In addition, DD is seen as having muscular peristalsis, which is good for distinguishing from the other abdominal cysts. Abdominal computerized tomography or magnetic resonance imaging is useful in representing lesions' anatomical structures including their location and size [1, 3, 5, 6]. Furthermore, magnetic resonance cholangiopancreatography (MRCP) or endoscopic retrograde cholangio-pancreatography (ERCP) can reveal associated pancreaticobiliary malformation [12, 15]. However, although imaging methods are helpful to the diagnosis, the most accurate diagnosis is made by histology.

Surgical management is the most common measurement for DD and is made in accordance with the relationship between the duplication and the pancreaticobiliary system. A total excision is an optimal approach. However, if the lesion occurs closed to the pancreaticobiliary tract, performing the total excision is difficult and risky. Recently, more and more cases which treated by endoscopic fenestration, have been reported and this method was thought to

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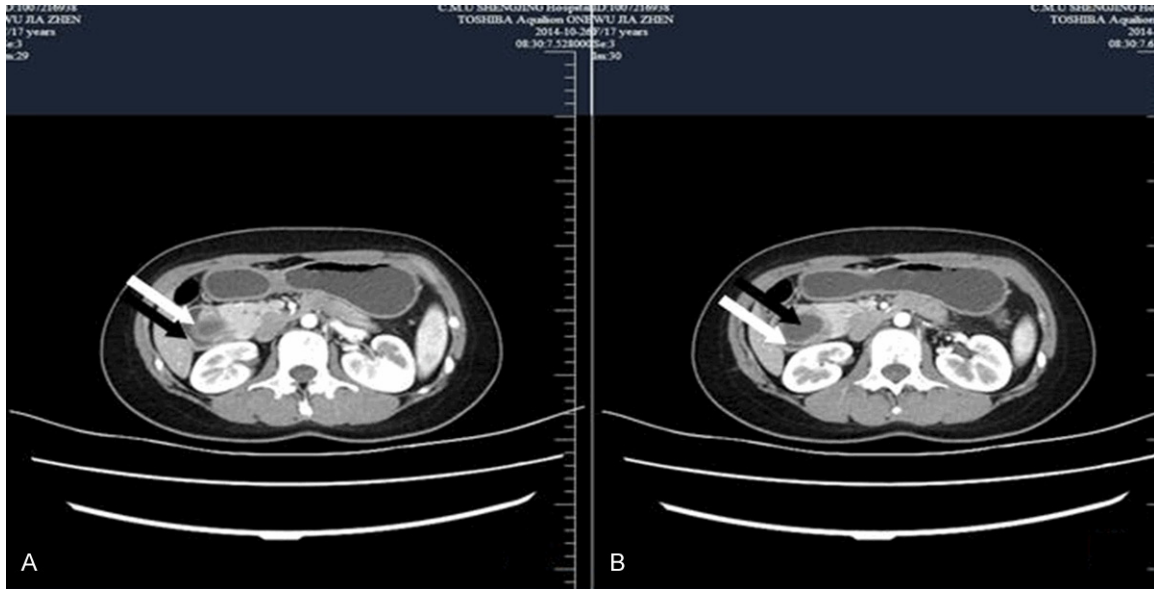


Figure 2. Upon abdominal contrast-enhanced computerized tomography, a cystic lesion of 3.4×3.3 cm in cross-section was found which located at the second portion of the duodenum (A), the cystic lesion's wall (white arrow) and duodenal wall (black arrow) was revealed by CT. It also demonstrated that the lateral wall of the second part of the duodenum was became thicker, and there was restricted liquid between tunica mucosa and tunica serosa of the lesion (B), the cystic lesion's cavity (black arrow) and duodenal cavity (white arrow) was found by CT.

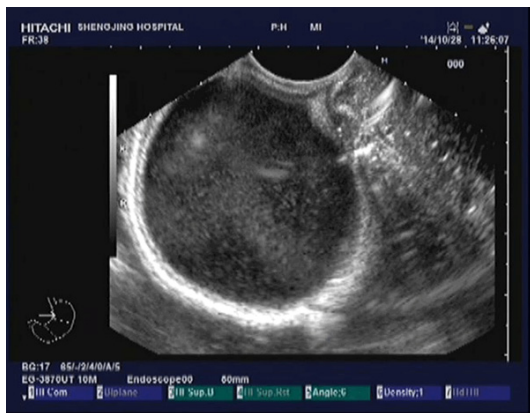


Figure 3. To find out the relationship between the cystic lesion and the biliary and pancreatic system, an endoscopic ultrasonography was done, which revealed no communication existed.

be safe and effective for intraluminal DD [16]. But, it has been reported that the DD has the possibility to become malignant [17]. In some cases, carcinoid tumor was found in the wall of the duplication cyst [18]. If only perform drainage endoscopically to relieve symptoms without excising the lesion, the duplication cyst may turn malignant. Therefore, we treated our patient with subtotal excision and internal derivation, which cured the patient.

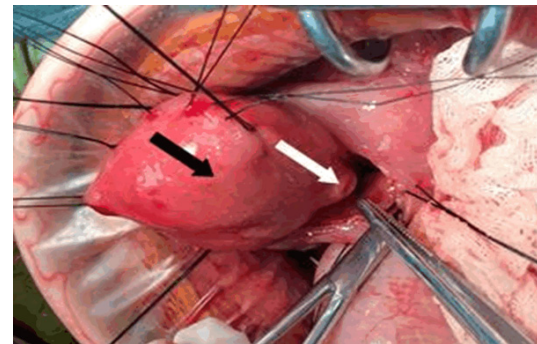


Figure 4. Operation was performed and we found an elliptic cystic lesion of 3.4×3.5 cm in diameter and anterolateral to the second part of the duodenum, the cystic lesion (black arrow) and duodenal papillae (white arrow) was found.

Conclusion

Duodenal Duplication (DD) is a rare congenital anomaly that is seldom reported in adolescents and its clinical manifestations are diverse. Here, we reported a very rare case of an adolescent who was diagnosed with non-communication cystic DD, presenting as abdominal pain and bowel obstruction. Imaging modalities were helpful to detect the lesions to make our diagnosis become more accurate, such as ultrasonography, CT imaging and MRI. Our patient

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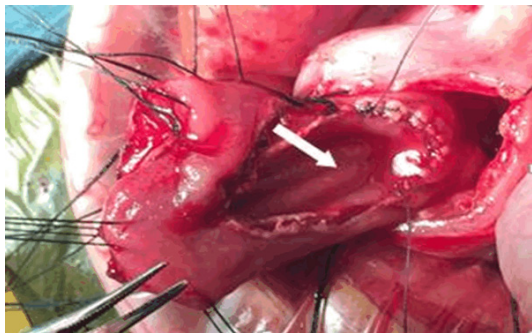


Figure 5. We performed the subtotal resection and the marsupialization (whit arrow) which was also called classical Gardner-Hart operation.

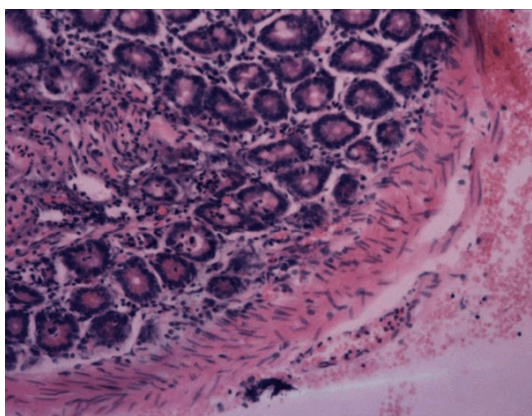


Figure 6. Histological findings revealed that duodenum-like mucosa, submucosa and muscle coats were found from the resected tissue.

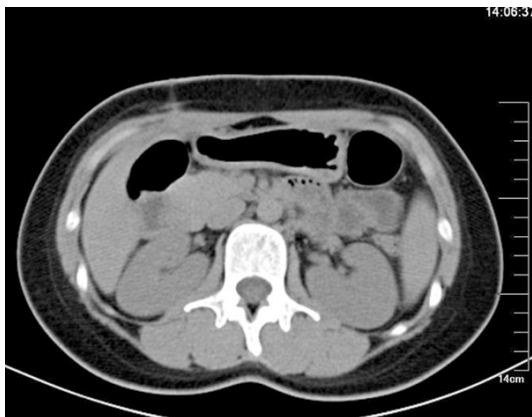


Figure 7. An abdominal CT was done in 3 months later which resulted for no abnormality.

underwent a successful subtotal excision and internal derivation. It is very important to be ware of duodenal duplication when evaluating a patient with a transient abdominal pain accompanied by duodenal obstruction.

Disclosure of conflict of interest

None.

Abbreviation

DD, Duodenal Duplication.

Address correspondence to: Dr. Ying Fan, Department of The Second General Surgery, Shengjing Hospital of China Medical University, #36 Sanhao Street, Heping District, Shenyang 110004, Liaoning Province, People's Republic of China. Tel: 086-24-96615-1-31211; E-mail: coolingpine78@163.com

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